Clinical Imaging 56 (2019) 47–51

Contents lists available at ScienceDirect

Clinical Imaging

journal homepage: www.elsevier.com/locate/clinimag

Body Imaging

Ovarian dermoid cyst complicated by small bowel obstruction, entero-ovarian fistula formation, and malignant degeneration

Yonah B. Esterson*, Marian Gaballah, Gregory M. Grimaldi, Micheal H. Raj, John S. Pellerito

Department of Radiology, Northwell Health System, The Donald and Barbara Zucker School of Medicine at Hofstra/Northwell, 300 Community Drive, Manhasset, NY 11030, USA

ARTICLE INFO

Keywords:
Dermoid
Teratoma
Mature cystic teratoma
Small bowel obstruction
Fistula
Malignant degeneration

ABSTRACT

Dermoid cysts, or mature cystic teratomas, are complicated by malignant degeneration in only 1–2% of cases. Rarely, dermoid cysts result in complications of small bowel obstruction or entero-ovarian fistula formation. In the current report we present the case of a 59-year-old female with a known dermoid cyst who presented with leukocytosis and was discovered to have a small bowel obstruction at the level of an ileo-ovarian fistula. The patient was taken to the operating room and was treated with total abdominal hysterectomy, bilateral salpingo-oopherectomy, and short segment small bowel resection. Review of the surgical pathology revealed areas of malignant degeneration within the dermoid cyst to squamous cell carcinoma. To our knowledge, this is only the second reported case of an ovarian dermoid cyst complicated by small bowel obstruction, entero-ovarian fistula formation, *and* malignant degeneration.

1. Introduction

Ovarian dermoid cysts, also known as mature cystic teratomas, are characterized pathologically by the presence of elements from at least two of three germ cell layers; ectoderm, mesoderm, and endoderm. Radiologists similarly identify dermoid cysts by their diverse contents, including fat, hair, and teeth [1].

Dermoid cysts are the most common germ cell neoplasm, typically affecting young adult females in their second and third decades of life. Although dermoid cysts are usually asymptomatic, approximately 20% of patients will experience complications including torsion, rupture, superimposed infection, and autoimmune hemolytic anemia [2,3]. Malignant degeneration is considered a rare complication of dermoid cysts, occurring in only 1–2% of cases, predominantly in middle-aged patients and those with larger dermoid cysts, most often to squamous cell carcinoma [3–6]. Even more rare, however, is the development of a small bowel obstruction caused by compression or invasion of the bowel by a dermoid cyst. Ovarian dermoid-related bowel obstruction has been reported in only seven prior cases [7–13]. Similarly, fistulization of ovarian dermoid cysts into an adjacent organ or viscus is reported to occur infrequently. In a review of 569 ovarian dermoid cases collected over a 27-year period, Stern et al. reported only one case of entero-ovarian fistula formation, a rate of only 0.2% [14]. Similarly, in a review of 1007 cases of dermoid cysts, Peterson et al. failed to document a single case of fistulization to an adjacent organ [15]. In the current report, we present only the second case in the literature in which these three uncommon complications (malignant degeneration, fistulization, and small bowel obstruction) are seen in one patient with an ovarian dermoid cyst [13].

2. Case report

A 59-year-old female scheduled for surgical resection of a dermoid cyst presented to pre-surgical testing services for pre-operative evaluation. When blood work revealed leukocytosis to 29.0 K/μL, the patient was asked to go directly to the emergency department (ED). In the ED, the patient denied fevers or chills. Other than her dermoid cyst, she had no relevant past medical or surgical history. Physical exam demonstrated a palpable abdominal mass, however the abdomen remained soft and non-tender. In the ED, the patient was normotensive but febrile to 101.1 °F and tachycardic to 106 beats per minute. Lab results were significant for leukocytosis to 30.2 K/μL, mild hyponatremia to 132 mmol/L, mild hypochloremia to 91 mmol/L, and hypoalbuminemia to 2.9 g/dL. A CT of the abdomen and pelvis demonstrated a 15.7 × 15.7 × 21.0 cm left adnexal mass containing fat and calcifications, consistent with a dermoid cyst (Figs. 1a, 1b, and 1d). A loop of ileum was inseparable from the anterior aspect of the mass, with communication demonstrated between the bowel lumen and the

* Corresponding author at: 300 Community Drive, Manhasset, NY 11030, USA.
E-mail addresses: YEsterson@northwell.edu (Y.B. Esterson), MGaballah@northwell.edu (M. Gaballah), GGrimald@northwell.edu (G.M. Grimaldi), MRaj@northwell.edu (M.H. Raj), JPelleri@northwell.edu (J.S. Pellerito).

https://doi.org/10.1016/j.clinimag.2019.03.005
Received 27 December 2018; Received in revised form 13 February 2019; Accepted 7 March 2019

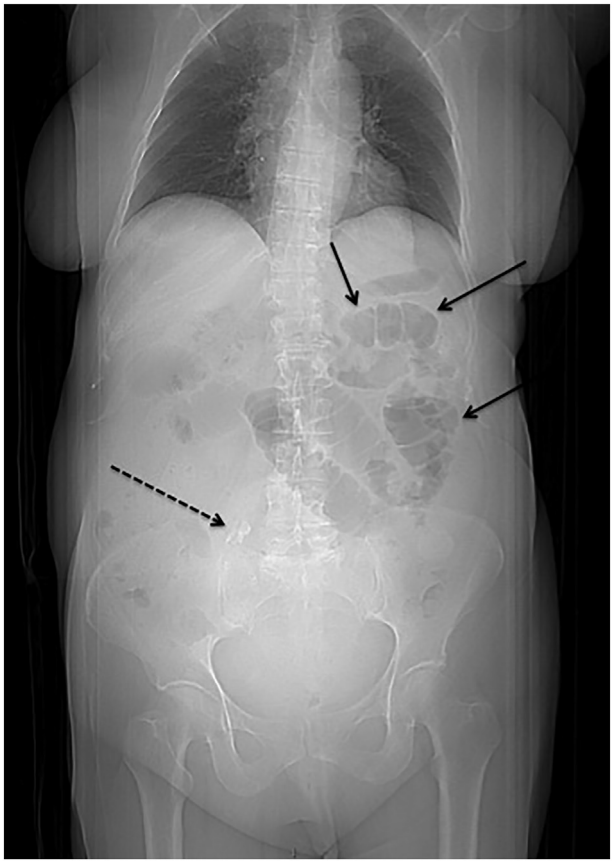

Fig. 1a. Scout CT image demonstrating dilated, gas-distended loops of bowel in the left hemiabdomen (solid black arrows) and coarse calcifications in the right lower quadrant (dashed black arrow).

Fig. 1b. Coronal CT image in soft tissue window demonstrating a heterogeneous lower abdominal mass (solid white arrows) containing fat (white asterisk) and coarse calcifications (dashed white arrow), compatible with a dermoid cyst. The mass is also noted to contain gas (dotted white arrow). Dilated loops of proximal small bowel are present in the right upper quadrant (solid black arrow), with collapsed loops of distal bowel in the right lower quadrant (curved solid white arrow).

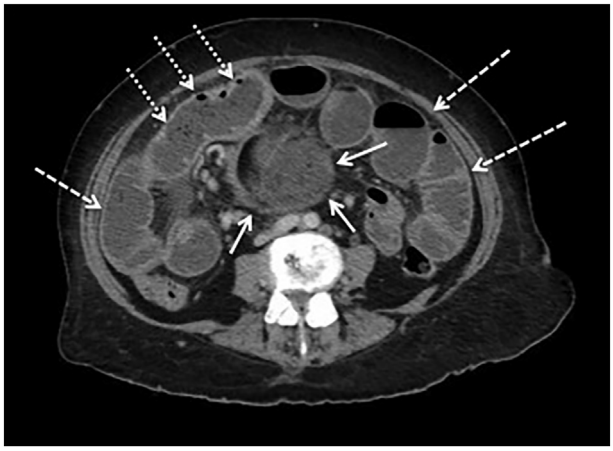

Fig. 1c. Axial CT image in soft tissue window demonstrating a fecalized loop of small bowel at the anterior aspect of the right hemiabdomen (dotted white arrows) with additional dilated loops of bowel present throughout the abdomen (dashed white arrows), suggesting the presence of a small bowel obstruction. The superior-most aspect of the patient's dermoid cyst is seen centrally (solid white arrows).

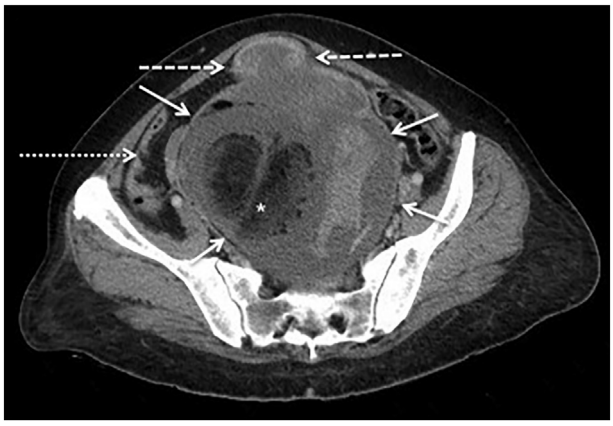

Fig. 1d. Axial CT image in soft tissue window demonstrating a heterogeneous pelvic mass (solid white arrows) with a fat component (asterisk). A loop of ileum is inseparable from the anterior aspect of the mass (dashed white arrows) and there is fistulous communication between the two structures, explaining the presence of gas within the mass demonstrated in Fig. 1b. Collapsed loops of distal small bowel are present in the right lower quadrant (dotted white arrow).

contents of the dermoid cyst (Fig. 1d). Fistulous communication with bowel was also evidenced by the presence of gas within the dermoid cyst (Fig. 1b). Additionally, there was dilation of the small bowel proximal to the site of fistulous communication and collapse of distal small bowel loops, compatible with small bowel obstruction at the site of the entero-ovarian fistula (Figs. 1a–1d).

Given these findings, gynecologic oncology determined that emergent surgical intervention was warranted. Exploratory laparotomy revealed an inflamed left ovarian mass containing sebaceous fluid and hair, characteristic of a dermoid cyst. The mass was adherent to the omentum, left pelvic sidewall, and posterior pelvic peritoneum, as well as to a 6 cm section of ileum, corresponding to the site of fistulization noted on preoperative CT. The small bowel was partially obstructed with transition point at the level of the entero-ovarian fistula. The involved segment of small bowel was resected along with the left ovary and dermoid cyst, which were sent for pathologic evaluation. Frozen sections confirmed the diagnosis of dermoid cyst but also demonstrated areas of malignant degeneration to squamous cell carcinoma. Hysterectomy, right salpingo-oophorectomy, omentectomy, and primary small bowel re-anastomosis were then performed prior to closure.

Table 1
History of patients with a small bowel obstruction caused by an ovarian teratoma.

Authors[a]	Age[b]	Entero-ovarian fistula	Malignant degeneration	Dermoid laterality	Greatest dimension[c]
Al-Harfoushi et al.	71	No	No	Right ovary	13.5 cm
Bhasin et al.	25	Yes	No	Right ovary	–
Gupta and Gupta	5	No	No	Right ovary	10.0 cm
Sundar et al.	30	No	No	Right ovary	8.0 cm
von-Walter and Nelken	25	Yes	No	Left ovary	10.0 cm
Wakelin et al.	38	Yes	No	Right ovary	7.0 cm
Yarmohammadi et al.	48	Yes	Yes[d]	Left ovary	10.3 cm
Present case	59	Yes	Yes[d]	Left ovary	21.0 cm

[a] See references 7–13.
[b] Age is reported in units of years.
[c] Greatest tumor dimension was not available in one of eight cases.
[d] Malignant degeneration to squamous cell carcinoma.

Table 2
History of patients with an entero-ovarian fistula caused by an ovarian teratoma.

Authors[a]	Age[b]	Bowel involved[c]	Malignant degeneration[d]	Dermoid laterality	Greatest dimension[e]
Bhasin et al.	25	I	No	Right ovary	–
Carter et al.	36	SC	No	Left ovary	8.0 cm
Cebesoy et al.	30	R	No	Left ovary	10.0 cm
Chauhan et al.	55	I	No	Right ovary	9.2 cm
Chong et al.	85	DC and SC	Yes[f]	Right ovary	–
Conway and Hanna	26	TC	No	Right ovary	–
Dandia	9	R	No	Left ovary	4.2 cm
Farkouh et al.	26	R	No	Right ovary	–
Figiel and Figiel	57	SC	Yes[f]	Right ovary	–
Federico et al.	31	R	No	Bilateral	–
Gardner	31	R	–	Left ovary	–
Goldenberg	41	SC	–	Left ovary	8.0 cm
Jalihal et al.	44	SC	No	Right ovary	8.3 cm
Khanna et al.	18	SC	No	Left ovary	8.0 cm
Kim et al.	17	R	No	Right ovary	11.0 cm
Kizaki et al.	43	R	No	Left ovary	11.0 cm
Landmann and Lewis	22	R	No	Right ovary	9.0 cm
Livesey et al.	70	R	No	Left ovary	7.0 cm
Lowe et al.	23	SC	–	Left ovary	7.0 cm
Mitui et al.	72	RSC and I	Yes[f]	Right ovary	8.0 cm
Sasaki et al.	48	RSC	No	Right ovary	5.0 cm
Shiels et al.	21	SC	No	Left ovary	7.0 cm
Singh et al.	23	RSC	No	Left ovary	5.0 cm
Song and Conner	73	SB	Yes[f]	Left ovary	24.0 cm
Stern et al.	29	I	No	Left ovary	13.0 cm
Thomas and Exley	45	SC, A, and I	–	Right ovary	–
von-Walter and Nelken	25	J and TC	No	Left ovary	10.0 cm
Wakelin et al.	38	I	No	Left ovary	7.0 cm
Yarmohammadi et al.	48	SB	Yes[f]	Left ovary	10.3 cm
Present case	59	I	Yes[f]	Left ovary	21.0 cm

[a] See references 2,9,11–14,21–42.
[b] Age is reported in units of years.
[c] J = jejunum, I = Ileum, SB = unspecified small bowel, A = appendix, TC = transverse colon, DC = descending colon, SC = sigmoid colon, RSC = rectosigmoid colon, R = rectum.
[d] Information on the presence or absence of malignant degeneration was not available in four of thirty cases.
[e] Greatest tumor dimension was not available in eight of thirty cases.
[f] Malignant degeneration to squamous cell carcinoma.

Table 3
History of patients with an ovarian teratoma complicated by small bowel obstruction, entero-ovarian fistula formation, *and* malignant degeneration.

Authors[a]	Age[b]	Malignancy harbored[c]	Bowel invaded[d]	Dermoid laterality	Greatest dimension
Yarmohammadi et al.	48	SCC	SB	Left ovary	10.3 cm
Present Case	59	SCC	I	Left ovary	21.0 cm

[a] See reference 13.
[b] Age is reported in units of years.
[c] SCC = squamous cell carcinoma.
[d] I = Ileum, SB = unspecified small bowel.

Final surgical pathology demonstrated poorly differentiated (grade G3) squamous cell carcinoma arising in a mature cystic teratoma with extension into the ileal serosa and mesentery. Surgical margins, as well as the uterus, bilateral fallopian tubes, and excised mesenteric lymph nodes were negative for malignancy.

3. Discussion

Primary ovarian tumors can be classified as surface epithelial-stromal tumors, sex cord-stromal tumors, or germ cell tumors. Germ cell tumors include yolk sac tumors, dysgerminomas, mixed germ cell tumors, embryonal carcinomas, choriocarcinomas, and teratomas. Teratomas are divided into mature cystic teratomas, mature solid teratomas, immature teratomas, and specialized teratomas [16]. Mature cystic teratomas, or dermoid cysts, are fairly common, constituting one-fifth of all ovarian tumors in the adult population and one-half of all ovarian tumors in the pediatric population, and are therefore frequently encountered by radiologists as an incidental finding at imaging of the female pelvis [17]. Given that CT and MRI are particularly sensitive for the presence of fat, the diagnosis of a mature cystic teratoma can be easily made in the 93% of mature cystic teratoma cases that contain fat [18]. On CT, the tumor will demonstrate a component of fat density, usually alongside soft tissue and calcium components. On MRI, the fat-containing component of the tumor will demonstrate T1 hyperintense signal that suppresses on T1-weighted chemical selective fat-saturated images. Chemical shift imaging can be helpful to detect microscopic fat in tumors containing only minimal fat [3]. Similarly, mature cystic teratomas have several characteristic appearances at ultrasound imaging. These include the presence of a dermoid plug, or Rokitansky nodule, a peripheral solid echogenic nodule with posterior acoustic shadowing projecting into the cystic cavity; an echogenic or partly echogenic mass due to its sebum and hair content; the presence of a dermoid mesh, innumerable thin echogenic lines suspended throughout the cyst cavity; and fluid-fluid levels formed by less dense echogenic sebum floating on more dense hypoechoic or anechoic aqueous material [19,20].

Small bowel obstruction is a rare complication of ovarian mature cystic teratomas, with only seven prior cases reported (Table 1) [7–13]. Fistulization is another rare complication of ovarian dermoid cysts. When they do fistulize, the urinary bladder is the most commonly affected organ [21]. Entero-ovarian fistulas secondary to dermoid cysts have been reported in only twenty-nine prior cases in the English language literature (Table 2) [2,9,11–14,19,21–42]. Proposed mechanisms of fistula formation include torsion of the tumor with resultant ischemia and pressure necrosis, infection with adhesion to and eventual rupture into an adjacent organ, and malignant transformation with direct invasion [37,39]. Of the thirty dermoids that had fistulized to bowel (inclusive of the current case), twenty fistulized to only large bowel, seven to only small bowel, and three to both small and large bowel. Interestingly, fistulization does not imply malignant degeneration. In fact, only six of the thirty dermoids that had fistulized to bowel (inclusive of the current case) demonstrated evidence of malignant degeneration at pathology. Instead, findings associated with malignant degeneration are reported to include patient age greater than 45 years old and tumor diameter greater than 9.9 cm [3]. Of the patients with malignant dermoid cysts referenced in Tables 1–3, all were older than 45 years old and only one had a tumor size less than 9.9 cm, although tumor size was not available in two of these cases.

To our knowledge, the current case is one of only two reported cases in which a bowel obstruction was caused by malignant degeneration of a mature cystic teratoma which had fistulized to the bowel (Table 3) [13]. Both cases underwent malignant degeneration to squamous cell carcinoma, both invaded the small bowel, both were larger than 9.9 cm, both originated from the left ovary, and both patients were older than 45 years of age.

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